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Chronic Pain in People with Physically Disabling Conditions: A Review of the Application of Biopsychosocial Models

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1. Introduction

There are a wide range of conditions which can result in physical disabilities, affecting more than a billion people or approximately 15% of the population worldwide (World Health Organisation, 2011). Disability is an umbrella term for the restrictions and impairments arising from the interaction between an individual with a health condition and the environment (World Health Organisation, 2011). The physical conditions from which disability arises are diverse and heterogeneous, but include both diseases, such as multiple sclerosis, as well as traumatic and non-traumatic injuries, including spinal cord injury and brain injuries. Research concerning the occurrence of chronic pain (defined as pain persisting beyond the period of healing, usually operationalized as three or six months) among people with physical disabilities, and the impact upon those individuals affected, is relatively limited compared to that focusing on primary pain conditions, such as low back pain. Within the available literature the focus is largely biomedical, with the majority of available research exploring biological factors and medical interventions. A great deal has been gained in the management of primary pain conditions through investigating psychological and social factors, and developing interventions such as cognitive behavioural pain management programs to target these factors. This chapter will describe the current understanding of the role of psychological and social factors in understanding the experience of chronic pain in the context of a physically disabling condition, and the use of interventions addressing these factors in this population.

2. The nature of chronic pain in people with physically disabling conditions

What is known about the nature of chronic pain among those with physically disabling conditions varies depending on the condition of interest. An examination of those publishing in the area suggests that often there are only a small number of groups involved in this research. Most of these research groups only work with one specific condition, with only a very small number working across a range of physically disabling conditions. As a result of these research silos, there are many inconsistencies in the approaches taken in investigating pain in the different conditions. Interpreting the findings of this body of work, particularly attempting to make comparisons across conditions, should therefore be done with caution keeping in mind some important caveats. First, there are variations across studies concerning the nature of the pain conditions which are the subject of investigation,

including the duration of the pain condition being explored. Some studies focus on chronic pain (with various criterion applied, commonly either three or six months duration), where others report on episodic, procedural or acute pain or do not specify which type of pain participants are experiencing. Second, some studies restrict their attention to pain thought to be specific to the physically disabling condition in question (such as headaches in those with traumatic brain injury), putting aside those deemed to be general (such as low back pain) which may be overlooked by teams specialising in a specific disabling condition rather than pain itself (Ivanhoe & Hartman, 2004). Finally, studies draw samples from a number of different populations including community samples, membership organisations for people with the physically disabling condition in question, and clinical services, including tertiary services for either pain or a specific physically disabling condition. There are also the usual methodological issues associated with the use of cross-sectional designs and the inevitable reliance upon self-report measures which is a hallmark of this area of research. Despite these issues, there is increasing evidence of a significant prevalence of chronic pain among those with physically disabling conditions.

2.1 Injury related conditions

Injury related conditions for which there is data regarding the nature of chronic pain includes those with a traumatic onset, such as traumatic brain injury and spinal cord injury, and those with a non-traumatic onset, such as cerebral palsy which is thought to be due to an injury to the developing brain. The kinds of traumatic events from which these injuries arise, such as motor vehicle accidents and sporting injuries, may mean that the individual has acquired multiple injuries which may complicate the assessment of the relationship between specific pain conditions and the physically disabling condition of interest.

2.1.1 Traumatic brain injury

Specific pain conditions of interest among those with traumatic brain injury include headaches of various types, but others noted include complex regional pain syndrome (CRPS), heterotopic ossification, and pain due to spasticity (Ivanhoe & Hartman, 2004; Nampiaparampil, 2008). A recent systematic review of chronic pain among those with traumatic brain injury identified 23 studies meeting their criteria, from which they estimated the overall prevalence of chronic pain in people with traumatic brain injury at 51.5% among civilians, with 57.8% reporting chronic headache (Nampiaparampil, 2008). An interesting aspect of findings among this group is that prevalence of chronic headaches differs depending on the severity of the traumatic brain injury (Lahz & Bryant, 1996; Nampiaparampil, 2008), with higher prevalence reported among those with mild traumatic brain injury. It has been observed that those with traumatic brain injury may have other traumatic injuries, and studies regarding the pain conditions in this poly-trauma group are very limited (Dobscha, et al., 2009). The assessment of pain in those with traumatic brain injury, particularly those with severe enough injuries to result in significant impairments to cognition, language or behaviour, may be challenging and so result in less accurate estimates than in some other groups with physically disabling conditions.

2.1.2 Spinal cord injury

Relative to other conditions, there have been a larger number of studies examining chronic pain among those with spinal cord injury. Classifying the pain conditions associated with

spinal cord injury has been undertaken by a number of groups, with more concordance among the groups in relation to some pain types than others, such as neuropathic pains (Cardenas, Felix, Cardenas, & Felix, 2009; Finnerup, Baastrup, & Jensen, 2009; Siddall, Yezierski, & Loeser, 2000). A recent systematic review of the prevalence of chronic pain of all causes among those with traumatic spinal cord injury have identified more than forty high quality studies from across the world, but the authors note that despite this there are many contradictions and unanswered questions about the nature of chronic pain in this group (Dijkers, Bryce, & Zanca, 2009). Prevalence rates of the studies included ranged from 26 to 98 percent, but the authors cautioned that the heterogeneity of the studies involved precluded calculation of an overall prevalence rate. It should be noted that co-morbid traumatic brain injury are not uncommon among this group, often within the mild range (Bradbury, et al., 2008). While spinal cord injury has been included in the section on traumatic injuries, some spinal cord injuries develop as a result of disease activity, such as cancer, which may mean the individual experiences other chronic pain conditions. As with many other physically disabling conditions, pain is only one of many troublesome complications. However, a recent study found that relative to others it is the most common and is closely associated with functioning (Jensen, Kuehn, Amtmann, & Cardenas, 2007).

2.1.3 Amputation

Studies of pain post-amputation are largely related to either upper or lower limb amputations. Most studies report on traumatically acquired amputations, although those related to vascular or other conditions are also relatively common. The most common type of pain problems reported in these studies is phantom limb pain and residual (or stump) pain, although it is also possible for people to develop musculoskeletal pain following amputation, for example in the back (Hammarlund, Carlström, Melchior, & Persson, 2011; Kooijman, Dijkstra, Geertzen, Elzinga, & van der Schans, 2000). Prevalence estimates suggest that phantom limb pain occurs in approximately 45 to 80% of people post-amputation, depending partly upon whether those with upper or lower limb amputations are considered, with rates of residual pain similarly high but varying (Behr, et al., 2009; Desmond & Maclachlan, 2010; Dijkstra, et al., 2002; Ehde, et al., 2000; Kooijman, et al., 2000; Schley, et al., 2008).

2.1.4 Cerebral palsy

In contrast to many of the other physically disabling conditions upon which this chapter focuses, research on cerebral palsy related pain is not confined to studies of adults. The common use of registries in a number of countries also means that the populations from which samples are drawn are more complete than in many other conditions. A French study of adults with cerebral palsy found that 75% reported pain of any sort (Gallien, et al., 2007). Musculoskeletal pain has been the subject of most investigation among this population. Prevalence estimates range from approximately one third to two thirds, with the pattern of distribution across the body different depending on the type of cerebral palsy (Engel, Jensen, Hoffman, & Kartin, 2003; Jahnsen, Villien, Aamodt, Stanghelle, & Holm, 2004; Schwartz, Engel, & Jensen, 1999; Vogtle, 2009). Although studies of children are more common in this group, they are limited by a number of factors regarding the measurement of pain and often rely on parental report. However, the available studies suggest 50 to 75 % of children are affected by pain, with approximately one third experiencing moderate to severe pain (Parkinson, Gibson, Dickinson, & Colver, 2010; Russo, Miller, Haan, Cameron, & Crotty, 2008).

2.2 Disease related conditions

Chronic pain has been investigated in the context of a range of diseases which are associated with physical disability. Some of the central pains, such as central post-stroke pain, associated with these conditions have only been recognized relatively recently, and have been subject to intense investigation.

2.2.1 Multiple sclerosis

A range of pain conditions, both general and specific, have been reported in people with multiple sclerosis. These include those related to spasm, neuropathic pains of various types (including trigeminal neuralgia and L'hermitte's sign), back pain and headaches. A recent systematic review of pain among people with multiple sclerosis found twenty-one studies reporting pain prevalence, with an overall range of 29 to 86%, and with few consistent findings related to the relationship between the report of pain and multiple sclerosis characteristics (O'Connor, Schwid, Herrmann, Markman, & Dworkin, 2008). Further, this study reported that between 11 and 23% of people reported pain as a symptom at the onset of their multiple sclerosis. Central pain, including trigeminal neuralgia, has been reported in approximately one third of people with MS (Osterberg, Boivie, & Thuomas, 2005).

2.2.2 Stroke

A number of chronic pain conditions are observed in people who have had a stroke, including shoulder pain, spasticity related pain and headaches, and central post-stroke pain is a uncommon condition particular to stroke. Estimates of the prevalence of chronic pain between studies are variable. Approximately 11 to 21% of people following a first stroke have been reported to have a stroke related pain condition up to 16 months following the stroke (Appelros, 2006; Jonsson, Lindgren, Hallstrom, Norrving, & Lindgren, 2006; Lundstrom, Smits, Terent, & Borg, 2009), whereas 42% of people following stroke attending an out-patient rehabilitation clinic had chronic musculoskeletal pain (Kong, Woon, & Yang, 2004). Neuropathic and central pain conditions occur at a significant level among those who have had a stroke, including estimates of complex regional pain syndrome in 15% of those undergoing inpatient post-stroke rehabilitation and 7% with central poststroke pain (Kitisomprayoonkul, Sungkapo, Taveemanoon, & Chaiwanichsiri, 2010; Klit, Finnerup, Andersen, & Jensen, 2011).

2.2.3 Parkinson's Disease

Relatively little is known about the prevalence and nature of pain in people with Parkinson's Disease, although clinically it is reported to be observed frequently (Beiske & Loge, 2009). Ford (2010) has classified pain problems associated with Parkinson's Disease in five categories: musculoskeletal pain; radicular or neuropathic pain; dystonia-related pain; akathitic discomfort; and central or primary pain. Overall estimates of the prevalence of pain in this group range between 40 and 83% (Beiske & Loge, 2009; Ford, 2010). Pain has been reported to be the first reported symptom of Parkinson's Disease among a significant minority of those who initially present with non-motor symptoms (O'Sullivan, et al., 2008).

2.2.4 Muscular dystrophy

Duchenne muscular dystrophy, myotonic muscular dystrophy, type I and facioscapulohumeral muscular dystrophy have all been found to be associated with chronic

pain (Engel, Kartin, Carter, Jensen, & Jaffe, 2009; Jensen, Abresch, Carter, & McDonald, 2005; Jensen, et al., 2008; Miro, et al., 2009) although there are limited studies with these groups. More than half of the respondents in these studies reported the presence of chronic pain, with pain in the back and legs most commonly reported but with a large number of sites reported to be involved.

2.3 Summary

Although not a comprehensive review of all of those physically disabling conditions which may result in the experience of chronic pain, the above summary suggests that chronic pain is a relatively common but perhaps neglected consequence of physically disabling conditions (Osterberg, et al., 2005). The specific causes and the nature of chronic pain experienced may differ between physically disabling conditions, but its high prevalence indicates that it is a legitimate concern for health practitioners and researchers. Awareness of the likelihood of pain is a necessary, although not sufficient, condition for the effective management of such pain. There is evidence that there are misconceptions among health professionals about the likely occurrence of pain in those with physically disabling conditions (Piwko, et al., 2007). Indeed, studies of people with pain associated with various conditions, including spinal cord injury, cerebral palsy, and multiple sclerosis, indicate that those affected perceive the available treatments and access to them to be inadequate (Cardenas, et al., 2009; Henwood & Ellis, 2004; Kennedy, Lude, & Taylor, 2006; Pollmann, Feneberg, & Erasmus, 2004; Wartan, Hamann, Wedley, & McColl, 1997). In addition, the economic costs of pain among those with physically disabling conditions appears to be large, with a Canadian study estimating that the cost of pain among people multiple sclerosis over a 6 month period was Can\$80 million (Piwko, et al., 2007).

3. Psychological and social factors in chronic pain among those with physically disabling conditions

Biopsychosocial models of pain, which are characterized by a focus on the interaction between biological, psychological and social variables in the pain experience, dominate the contemporary understanding of primary pain conditions, such as headache and low back pain (Gatchel, Peng, Peters, Fuchs, & Turk, 2007). The use of biopsychosocial models has also been promoted as the most appropriate framework for the understanding and management of disability of all types (World Health Organisation, 2011). The importance of these models is that they broaden the focus of health professionals to consider psychological and social factors which affect the course of a chronic pain condition. Crucially, where many chronic pain conditions were resistant to the available medical interventions, resulting in a significant proportion of the population living with chronic pain, biopsychosocial models offered a new treatment target focused on reducing the psychological distress and functional disability experienced by those with persisting pain (Blyth, et al., 2001).

Within the broad category of biopsychosocial models, those informed by cognitive behavioural theories are most common. These models particularly address the role of cognitions and behaviours in the development and maintenance of pain, as well as associated pain-related distress and disability. A wide range of cognitive and behavioural constructs have been investigated to determine their relationship with the experience of pain and associated disability and distress (Vlaeyen & Linton, 2000). There is some support for many of these constructs in individual studies. However, among these constructs, pain

catastrophizing, pain self-efficacy, and avoidance have all been found to be consistently related to the experience of pain and associated disability or distress across different populations and methodologies. In addition, the presence of altered mood, such as symptoms of depression and anxiety, or the co-existence of psychiatric disorders such as post-traumatic stress disorder, all seem to be important in influencing the course of a chronic pain condition.

The application of biopsychosocial models to chronic pain among those with physically disabling conditions has lagged significantly behind its use among primary pain conditions, where it has long been acknowledged that presence of pain or the intensity of pain does not fully explain pain-related disability or distress (Heneweer, et al., 2007). Among the physically disabling conditions outlined above with documented, significant rates of associated chronic pain there were no peer-reviewed publications available addressing psychosocial variables in the case of Parkinson's Disease. In addition, the investigation of variables has been patchy across the different conditions, and there are few specific models proposed for those with chronic pain in the context of physically disabling conditions. Some of the reasons for this are unclear, although it may be suggested that the obvious presence of pathology among those with physically disabling conditions results in a tendency to discount the possible role of other factors in causing pain-related disability or distress. This is despite repeated findings across a number of diagnoses that condition-related variables, such as severity of injury or illness, are frequently not at all or only weakly related to pain intensity and related disability or distress (Hoffman, et al., 2007).

It is notable that while biopsychosocial models do not dictate that only negative or adverse outcomes are possible following development of pain, much of the available research focuses on factors associated with poor outcomes. In contrast, a study of people with spinal cord injuries or multiple traumas examined factors which differentiated different adjustment trajectories and identified three: the resilience trajectory, characterized by low levels of mental health symptoms at both early and late stage of admission following injury, the recovery trajectory where the individual shows an improving pattern of mental health symptoms, and the distress trajectory, where higher levels of mental health symptoms at the early stage are sustained in the longer term (Quale & Schanke, 2010). The study reports that the latter accounted for only one fifth of the participants in their study, and that maximum pain at admission differentiated those in the resilience vs the distress trajectory.

Studies investigating the relationship between psychosocial factors and chronic pain in people with physically disabling conditions fall into two main categories. First, psychosocial factors are examined as predictors of chronic pain or pain-related disability, for example does a particular way of thinking about pain have an impact on how much pain is experienced. Second, chronic pain is examined as a contributor to adjustment following onset of a physically disabling condition, for example do those with chronic pain as a result of a physically disabling condition have higher levels of depressive symptomatology. The rapid development of theory and research related to biopsychosocial models of chronic pain has led to some overlap of concepts. A good example of this being that pain catastrophizing first appeared as one of many unhelpful coping strategies, but has now been reframed as a belief alongside others such as self-efficacy or helplessness. It is therefore somewhat difficult to categorize the current literature into particular themes. The purpose of this section is to describe the current evidence regarding the relationship between psychological and social factors and chronic pain in people with physically disabling conditions, and to identify gaps in the current literature which require further investigation.

3.1 Psychological factors

Under the category psychological factors, the main variables to consider are mood and mental health, cognitive responses to pain, and behavioural responses to pain. The extent of the literature in these three areas varies markedly across the various physically disabling conditions under consideration.

3.1.1 Mood and mental health

The association between chronic pain and changes in mood and mental health, including symptoms of depression and anxiety, is perhaps the most frequently explored aspect of biopsychosocial models among those with physically disabling conditions. In some physically disabling conditions, a clear relationship between chronic pain and psychological distress has been consistently demonstrated, whereas in others the findings are more mixed. Pain has been found to be associated with psychological distress in most of the studies identified in cerebral palsy, traumatic brain injury, multiple sclerosis, amputation, spinal cord injury, and muscular dystrophy (Engel, et al., 2003; Engel, Schwartz, Jensen, & Johnson, 2000; Hoffman, et al., 2007; Kalia & O'Connor, 2005; Kratz, et al., 2010; Middleton, Tran, & Craig, 2007; Miro, et al., 2009; Motl, McAuley, Snook, & Gliottoni, 2009; Nicholson Perry, Nicholas, & Middleton, 2009; Nicholson Perry, Nicholas, Middleton, & Siddall, 2009; Norrbrink Budh, Hultling, & Lundeberg, 2005; Stormer, et al., 1997; Turner, Jensen, Warm, & Cardenas, 2002), although the strength of the relationship has varied across studies and conditions. In addition, studies in people with spinal cord injury pain found that continuous pain, as opposed to intermittent pain, was associated with higher levels of depression and anxiety, and conversely more stress among women with spinal cord injuries was associated with consistent reports of pain over a ten year period (Norrbrink Budh & Osteraker, 2007; Rintala, Hart, & Priebe, 2004). Negative mood has also been reported to be a trigger to exacerbations in chronic pain among people with spinal cord injury (Widerstrom-Noga & Turk, 2004). Some studies have presented exceptions to this general rule in the case of cerebral palsy, multiple sclerosis and stroke (Hirsh, Gallegos, Gertz, Engel, & Jensen, 2010; Kong, et al., 2004; Newland, Naismith, & Ullione, 2009; Newland, Wipke-Tevis, Williams, Rantz, & Petroski, 2005). Moderators of this relationship include gender, with the relationship not being found in males with multiple sclerosis in one study, and aetiology (traumatic versus non-traumatic) for amputation in one study moderating the relationship at early time points (Kalia & O'Connor, 2005; Kratz, et al., 2010). Physically disabling conditions in which findings are mixed in this regard include those with phantom limb pain and stump pain following amputation (Fisher & Hanspal, 1998). While most of these studies have been conducted with adults with physically disabling conditions, a study with a large sample of older children with cerebral palsy suggests that children with moderate or severe pain are significantly more likely to have higher levels emotional and behavioural problems (Parkes, et al., 2008). One study in people with multiple sclerosis found that affective memory biases, a measure of vulnerability to depression, may mediate the relationship between chronic pain and depressive symptoms in this group (Bruce, Polen, & Arnett, 2007).

Most studies of the relationship between pain and depression in physically disabling conditions solely report on cross-sectional associations. In some studies, however, they specifically examine pain as a predictor of depression, or in other cases the reverse. Determining the direction of the relationship have proved problematic, although there is some evidence among people with spinal cord injury to support the hypothesis than persisting pain

is a driver of depression rather than the converse (Cairns, Adkins, & Scott, 1996; Putzke, Richards, Hicken, & DeVivo, 2002). The presence of depression at one time point has been reported to be a risk factor for pain at a later time point among those with multiple sclerosis and spinal cord injury (Buchanan, Wang, Tai-Seale, & Ju, 2003; Putzke, et al., 2002).

Depression is associated with pain-related interference in a number of physically disabling conditions, including amputation, multiple sclerosis and spinal cord injury (Kratz, et al., 2010; Nicholson Perry, Nicholas, & Middleton, 2009; Nicholson Perry, Nicholas, Middleton, et al., 2009; Norrbrink Budh, et al., 2005; Norrbrink Budh & Osteraker, 2007; Osborne, et al., 2006; Turner, et al., 2002). There is evidence of a similar moderating effect of depression upon the relationship between pain and disability among those with spinal cord injury as is seen in other chronic pain populations (Borsbo, Peolsson, & Gerdle, 2009), and a similar but less clear interaction between these variables in those with traumatic brain injury (Hoffman, et al., 2007). The impairment to quality of life attributable to chronic pain has been reported to be related to depressive symptoms among individuals with spinal cord injury (Cruz-Almeida, Alameda, & Widerstrom-Noga, 2009). In addition, negative moods, boredom and stress reported in a large sample of older children with cerebral palsy was found to be significantly predicted by the presence of pain, although only to contribute a relatively small proportion of variation in this aspects of quality of life and interestingly overall quality of life was found to be consistent with other children without cerebral palsy (Dickinson, et al., 2007).

Studies of the relationship between chronic pain and anxiety among people with physically disabling conditions are less common. However, in studies of people with multiple sclerosis, anxiety has been found to be positively associated with pain severity, particularly among women (Kalia & O'Connor, 2005; Motl, et al., 2009). Studies among people with spinal cord injury have also shown a significant relationship between anxiety and pain severity (Nicholson Perry, Nicholas, & Middleton, 2009; Nicholson Perry, Nicholas, Middleton, et al., 2009; Norrbrink Budh, et al., 2005; Norrbrink Budh & Osteraker, 2007). There are a few studies examining the relationship between pain and post-traumatic stress disorder (PTSD). In a study of people with both traumatic and non-traumatic amputation, pain and pain-related interference was positively correlated with PTSD symptoms in both groups (Kratz, et al., 2010). Pain-related anxiety, often measured as a combination of cognitions, behaviours and emotion, has also been found to moderate the relationship between chronic pain and disability among those with spinal cord injury, with those reporting higher levels of pain related anxiety experiencing greater disability (Borsbo, et al., 2009). Anger has been less well investigated, although it has been shown to moderate the perception of pain in people with spinal cord injury (Conant, 1998; Summers, Rapoff, Varghese, Porter, & Palmer, 1991).

3.1.2 Cognitive responses to pain

The relationship between cognitive responses to pain, or beliefs, and pain-related disability and distress has been explored in those with a number of the physically disabling conditions of interest. Pain catastrophizing, characterized by a tendency to negative and unrealistic beliefs in response to pain, and is the cognitive factor with the greatest body of evidence supporting its role. While pain catastrophizing has often been measured in questionnaires designed to measure coping strategies, it is best considered alongside other beliefs, and so will be reported on in this section.

Pain catastrophizing has been found to be associated with pain intensity among people with chronic phantom limb pain post-amputation, multiple sclerosis and spinal cord injury (Hill, Niven, & Knussen, 1995; Nicholson Perry, Nicholas, & Middleton, 2009; Osborne, Jensen,

Ehde, Hanley, & Kraft, 2007; Turner, et al., 2002; Vase, et al., 2011; Wollaars, Post, van Asbeck, & Brand, 2007). In a study of people with phantom limb pain, pain catastrophizing was also shown to significantly contribute to wind-up-like pain when anxiety and depression were controlled for (Vase, et al., 2011)

Pain catastrophizing has been found to be positively associated with pain-related disability among those with spinal cord injury, cerebral palsy, phantom limb pain, muscular dystrophy and multiple sclerosis (Borsbo, et al., 2009; Douglas, Wollin, & Windsor, 2008; Engel, et al., 2000; Hill, et al., 1995; Miro, et al., 2009; Molton, et al., 2009; Nicholson Perry, Nicholas, & Middleton, 2009; Nicholson Perry, Nicholas, Middleton, et al., 2009; Osborne, et al., 2007). Psychological functioning among people with spinal cord injury, multiple sclerosis, phantom limb pain, muscular dystrophy and cerebral palsy has been found to be negatively associated with pain catastrophizing (Douglas, et al., 2008; Engel, Jensen, & Schwartz, 2006; Engel, et al., 2000; Hanley, et al., 2004; Hill, et al., 1995; Miro, et al., 2009; Molton, et al., 2009; Nicholson Perry, Nicholas, & Middleton, 2009; Nicholson Perry, Nicholas, Middleton, et al., 2009; Osborne, et al., 2007; Smedema, Catalano, & Ebener, 2011; Ullrich, Jensen, Loeser, & Cardenas, 2007; Wollaars, et al., 2007). Pain catastrophizing has also been shown to mediate the relationship between pain severity and psychological distress and pain-related disability among people with spinal cord injury (Ullrich, et al., 2007). It has been suggested that pain catastrophizing may, in fact, be a function of disturbed mood. This suggestion is brought into question by findings in both phantom limb pain and spinal cord injury related chronic pain which shows that pain catastrophizing is associated with pain intensity when mood is controlled for (Ullrich, et al., 2007; Vase, et al., 2011). Among the many studies of individuals with spinal cord injury pain, veterans with the condition appear to have higher levels of pain catastrophizing than non-veterans (Ullrich, Jensen, Loeser, Cardenas, & Weaver, 2008).

While most of these studies are cross-sectional in nature, a prospective study of people with spinal cord injury with chronic pain found that over a six month period decreases in pain catastrophizing were associated with decreased pain interference and improved psychological functioning (Hanley, Raichle, Jensen, & Cardenas, 2008). Conversely, a similar study in phantom limb pain found that pain catastrophizing at one month following amputation (that is, before chronic pain had developed) was predictive of decreased depressive symptoms and pain-related interference at both 12 and 24 months (Hanley, et al., 2004). While this may appear counter-intuitive, the authors suggest that the function of pain catastrophizing soon after amputation may be different to that in those with established chronic pain, who are the subject of most other studies on the subject.

Perceived control over pain has also been investigated, and there is less extensive evidence to support its role in relation to psychological functioning and disability in those with chronic pain secondary to physically disabling conditions. A study of people with spinal cord injury related chronic pain found that increases in perceived control over pain in a six month period was related to decreased pain intensity and pain interference, as well as increased psychological functioning, although the former was a non-significant finding (Hanley, et al., 2008). External locus of control in relation to pain as also been positively associated with depression among people with spinal cord injury related pain (Wollaars, et al., 2007). In addition, two studies of people with phantom limb pain, including a prospective study of people with phantom limb pain from one to 24 months, demonstrated some weak evidence for its influence on pain intensity, psychological functioning, and pain-related disability (Hanley, et al., 2004; Hill, et al., 1995).

Other findings related to pain-related beliefs have also been noted but with much less consistency. A belief that pain is constant or enduring has been found to significantly predict both pain intensity and interference to activities due to pain among those with multiple sclerosis (Douglas, et al., 2008). The lower endorsement of the belief that others should be solicitous in response to pain behaviours was associated with better psychological functioning among people with muscular dystrophy (Miro, et al., 2009)

3.1.3 Behavioural responses to pain

Comparison of the use of behavioural responses to pain, also commonly referred to as coping strategies, among people with chronic pain secondary to physically disabling conditions to those with chronic primary pain conditions has revealed both similarities and differences. In people with cerebral palsy, use of behavioural coping strategies such as guarding and rest was reported to be less common and task persistence more common (Engel, et al., 2000). Conversely, cognitive coping strategies, such as diverting attention, reinterpreting sensations, and praying and hoping, were reportedly used more commonly. The authors suggest that some of these differences may be attributable to different background levels of the use of behavioural strategies such as resting and guarding, which may already be employed for non-pain related reasons among those with physically disabling conditions, and increased reliance on cognitive strategies over which they may be hypothesized to have more control.

The association of particular coping strategies with pain intensity or associated psychological distress or disability has been explored in a number of physically disabling conditions, including spinal cord injury, but with few significant relationships detected (Hanley, et al., 2008; Turner, et al., 2002). In some other studies, however, significant relationships have been found. In a study of people with phantom limb pain, behavioural activity was found to be associated with higher levels of pain, in contrast with findings in those with chronic primary pain conditions (Hill, et al., 1995). Passive coping strategies, including guarding, resting, asking for assistance, seeking social support and pacing, were found to be predictive of pain interference but not psychological functioning among people with spinal cord injury and muscular dystrophy (Miro, et al., 2009; Molton, et al., 2009). Reduction of activity, through resting or avoidance, has been associated with positively associated with pain interference in people with cerebral palsy and muscular dystrophy, and with symptoms of depression in people with cerebral palsy (Engel, et al., 2000; Miro, et al., 2009).

Seeking social support has been found to be positively associated with pain-related disability among people with cerebral palsy and muscular dystrophy (Engel, et al., 2006; Miro, et al., 2009), a finding that may initially appear counter-intuitive. The authors of the cerebral palsy study identified the fact that the items on the scale potentially reflect both adaptive and maladaptive aspects of social support seeking (Engel, et al., 2006). However, operant models of chronic pain suggest that pain contingent social support would result in increased disability, which may also provide a parsimonious explanation of the findings.

The extent to which respondents with multiple sclerosis believed they were able to control or decrease their pain through use of their coping strategies has been found to be associated with decreased pain intensity, however no specific coping strategy was predictive (Douglas, et al., 2008). Further, in the same study no coping strategy was found to be predictive of life interference due to pain and only coping by increasing activities was found to be associated with improved psychological functioning.

3.2 Social factors

Although clearly identified as part of the various biopsychosocial models of pain proposed, social factors have been relatively less well represented in the literature. Studies examining social factors most often report on perceived social support and partner responses to pain behaviours.

3.2.1 Social support

Studies examining the associations between social support and pain are available in people with limb loss, spinal cord injury, multiple sclerosis and muscular dystrophy.

Social support was found to be negatively associated with pain in studies among people with traumatic limb loss, whereas no relationship was found in people with spinal cord injury pain (Kratz, et al., 2010; Stroud, Turner, Jensen, & Cardenas, 2006). Studies of people with multiple sclerosis have resulted in mixed findings, with negative associations with pain in one study and no association in the other (Motl, et al., 2009; Osborne, et al., 2007). A study designed to identify factors which were predictive of consistency of pain over ten years among people with spinal cord injury found that among male respondents, receiving less social support during the first phase of the study was predictive of continuing pain over the life of the study (Rintala, et al., 2004)

Associations between lower levels of social support and greater pain-related disability has been found in people with non-traumatic limb loss, multiple sclerosis and muscular dystrophy, but not in people with spinal cord injury (Kratz, et al., 2010; Miro, et al., 2009; Motl, et al., 2009; Osborne, et al., 2007; Stroud, et al., 2006). In one study, greater social support at one month post amputation was predictive of greater reduction in pain interference at 12 and 24 months (Hanley, et al., 2004). In addition, increased social support has been found to be associated with lower levels of anxiety and depression in people with multiple sclerosis and with depression in people with spinal cord injury (Motl, et al., 2009; Stroud, et al., 2006). Among people with muscular dystrophy, social support was associated positively with psychological functioning (Miro, et al., 2009). Hanley et al. (2004) also looked at the relationship between social constraint, which is the need to hide one's feelings about the amputation from others, and pain and pain-related interference. Increased need for social constraint was associated with pain intensity and interference in both those with traumatic and non-traumatic limb loss in the 6 to 12 month period.

3.2.2 Partner responses to pain behaviours

The most common maladaptive form of partner response to pain behaviours reported in the general chronic pain literature is that of solicitous responding, which is a key mechanism in operant models of pain, hypothesized to increase pain-related disability. A study in people with spinal cord injury chronic pain found perceived solicitous responding from partners were unrelated to pain intensity, pain-related disability, or depression (Stroud, et al., 2006). One study in people with limb loss found that less frequent solicitous responding at one month post amputation was predictive of greater reductions in pain interference at 12 and 24 months (Hanley, et al., 2004). Other forms of partner responses to pain behaviour measured in people with spinal cord injury related chronic pain are negative and distracting responses (Stroud, et al., 2006). Negative responses, such as criticism, and distracting responses were both associated with higher depression, but not pain intensity or pain related disability. Most studies which report upon partner responses to pain behaviours in the context of physically disabling conditions provide information about participants

perception of their significant others solicitous responses to their pain behaviours. One study in people with spinal cord injury reported on partner's ratings of their own responses to pain behaviour, and it was notable that these were unrelated to pain intensity, depression or pain-related disability in their partner (Stroud, et al., 2006).

3.3 Summary

Reviewing the evidence presented here, a number of issues are apparent. First, that there is a great deal of variation across physically disabling conditions in the extent to which biopsychosocial factors have been investigated. Second, that over all the conditions considered, social factors are relatively less well explored and this remains a significant omission in the literature. Despite this, and the variation in the patterns in each specific physically disabling condition, across the majority of the conditions for which data is available it is clear that there is evidence to suggest that psychological and social factors are broadly related to pain intensity, as well as associated disability and distress. Across all the factors which have been investigated thus far, it appears that the findings related to pain catastrophizing and its association with pain intensity and related disability and distress are the most consistent. This suggests that pain catastrophizing should be explored as part of assessment protocols for people with chronic pain associated with physically disabling conditions. Further research in the area is clearly needed, particularly prospective studies that begin prior to the development of chronic pain, and which are sufficiently large to permit demographic and medical factors to be controlled for in the analyses.

4. Interventions focused on psychological and social factors

There are two major reasons why psychosocial interventions for pain might be considered for people with chronic pain secondary to physically disabling conditions. The first, that psychosocial variables are important contributors to variance in pain itself, as well as pain related distress or disability, and the second, that existing interventions based on biomedical models of pain are insufficient. The evidence presented in the previous section suggests that, while there are gaps in the literature, there is sufficient reason to think that psychosocial variables do make a significant contribution to pain and associated distress and disability. In addition, there is evidence that people with chronic pain secondary to a range of physically disabling conditions, including cerebral palsy, stroke and multiple sclerosis, are unlikely to be receiving treatment for their pain, are dissatisfied with the pain treatment available to them or report limited improvement in pain despite treatment (Engel, et al., 2003; Hirsh, et al., 2010; Kalia & O'Connor, 2005; Kong, et al., 2004).

Psychosocial interventions for people with chronic pain, predominantly behavioural and cognitive behavioural in origin, are well-established and supported by an extensive evidence base (Meldrum, 2007). A series of systematic reviews and meta-analyses attest to the efficacy of these programs among child and adults with primary chronic pain conditions, as well as early interventions designed to reduce the development of pain-related disability (Eccleston, Morley, Williams, Yorke, & Mastroiannopoulou, 2002; Eccleston, Yorke, Morley, Williams, & Mastroiannopoulou, 2003; Linton & Nordin, 2006; Morley, Eccleston, & Williams, 1999).

4.1 The nature of psychosocial interventions

A very small number of studies have been published that specifically report on the use of psychosocial interventions among people with physically disabling conditions. The majority

of these have involved group-based cognitive behavioural pain management programs, but they also include cognitive restructuring and hypnosis. The potential use of such interventions in people with spinal cord injury was identified in the early 1990's (Umlauf, 1992), but a review of the literature concerning the application and evaluation of these programs among any group with a physically disabling condition reveals a disappointingly small number of studies and little translation into standard practice.

4.2 Feasibility and acceptability of psychosocial interventions

A study undertaken in the US specifically examined the issue of the feasibility and acceptability of psychosocial interventions, with a mixed sample of individuals with chronic pain of more than six months duration occurring secondary to multiple sclerosis, amputations, spinal cord injury and cerebral palsy (Ehde & Jensen, 2004). The study found that both the cognitive restructuring intervention, and the control condition which was an educational intervention, were both rated positively by the participants. A study of a cognitive behavioural pain management program for people with spinal cord injury neuropathic pain reported that attendance at the group was high and participants reported that they were very satisfied with the program (Norrbrink Budh, Kowalski, & Lundeborg, 2006). Authors of another study, examining the effectiveness of a cognitive behavioural pain management program for people with spinal cord injury, provide an analysis of the issues encountered in the implementation of the program (Nicholson Perry, Nicholas, & Middleton, 2010; Nicholson Perry, Nicholas, & Middleton, 2011). These findings suggest that these interventions are potentially acceptable, at least to people with spinal cord injury related pain.

4.3 Use and effectiveness of psychosocial interventions

Psychosocial interventions for pain either described for use with or evaluated with people with physically disabling conditions are extremely few. They are mainly cognitive behavioural, group-based pain management programs, but examples of the use of cognitive restructuring alone and hypnosis are also reported.

4.3.1 Cognitive behavioural group-based pain management programs

Four group-based, cognitive behavioural pain management program of various sorts are described in the literature (Cundiff, Blair, & Puckett, 1995; Girona, 2004; Nicholson Perry, et al., 2010; Norrbrink Budh, et al., 2006). The main components of such interventions are represented in Table 1. The earliest reports in the literature of cognitive behavioural pain management programs in physically disabling conditions were for spinal cord injury pain and were descriptive. Cundiff and colleagues (1995) described the development of a group-based cognitive behavioural pain management program for people with spinal cord injury pain of all types. This involved many of the common components of pain management programs for primary pain diagnoses, including: the explanation of the self-management model, relaxation (including diaphragmatic breathing, guided imagery), the role of self-talk, and pain behaviours and their impact. Girona (2004) reported on an intervention which was characterized as an interdisciplinary pain management program for spinal cord injury shoulder pain. It was described as a functional preservation approach aimed at enabling individuals to maintain and improve functional capacities where injuries had already been sustained. The program was provided during a two week in-patient stay and comprised of:

medication adjustment; an exercise regimen designed to increase range of motion, endurance and stretch in upper limbs; biomechanical education; a psychoeducational component designed to enhance understanding of the self-management approach, promote problem-solving and implementation of strategies at home, raise awareness of compensatory responses that may be impacting upon psychological or physical well-being; and recreation therapy to encourage return to social and leisure interests. Preliminary data from eight participants in the program suggested improvements across a range of domains, including mood, sleep and pain intensity during shoulder range of motion testing.

Component	Description
Education regarding pain	Information is presented about the underlying pain mechanisms relevant to chronic pain, including central sensitization, as well as the limitations of medical treatment for chronic pain.
Goal-setting	Collaborative goal-setting related to a variety of goals across a wide spread of domains, including physical activities or mood, emphasizing the identification of short-term goals building towards long-term goals that are challenging but achievable in order to increase a sense of mastery.
Activity pacing	Adoption of quota or time based activities, systematically upgraded over time and linked to goals.
Relaxation	Applied relaxation to reduce muscle tension and improve sleep.
Functional exercise	Whole body reconditioning exercise programme related functional physical goals.
Stretch	Whole body daily stretch programme.
Cognitive therapy	Identification and modification of unhelpful thoughts regarding pain, such as catastrophizing.
Medication reduction	Gradual reduction of inappropriate or excessive pain medications using an agreed schedule.
Flare-up management & relapse prevention	Development of a plan to manage temporary increases in pain (flare ups) or other situations likely to trigger relapse.

Table 1. Common components of cognitive behavioural pain management programs

Two controlled studies have been published which have examined the effectiveness of cognitive behavioural group pain management programs for people with physically disabling conditions, both in those with spinal cord injury (Nicholson Perry, et al., 2010; Norrbrink Budh, et al., 2006). The first controlled study in the literature described a cognitive behavioural pain management program for people with neuropathic pain arising from a spinal cord injury (Norrbrink Budh et al., 2006). The program developed was very like the pain management programs described for people with primary pain diagnoses in content, although of shorter duration (totalling 50 hours over ten weeks). Compared with those in the no-treatment control group, those participating in the program showed significant improvements in depression and sense of coherence (a concept comprising comprehensibility, manageability and meaningfulness of the injury) over 12 months. While there were no other significant differences between the groups, the treatment group showed improvements in anxiety symptoms, emotional reaction and sleep from baseline to the 12-month evaluation, but no significant changes over time were observed in the other outcome

measures (including pain intensity and unpleasantness, health-related quality of life and life satisfaction). An Australian study compared a cognitive behavioural pain management program with standard care in a tertiary pain management service in Australia. The program was a modification of an existing program, the design and implementation of which is reported in depth elsewhere, which was approximately half of the usual number of contact hours (Nicholson Perry, et al., 2011). The group attending the pain management program showed an overall improved mood and pain-related disability at the end of the program compared with the controlled group. This was associated with significant decreases in pain catastrophizing and anxiety in the pain management program group. Three-quarters of people completing the pain management program reported a clinically significant improvement, in contrast to less than a third in the usual care comparison group, however long-term follow up data in this group suggested that benefits were not maintained at six months. Both programs were approximately half of the optimal dose (100 hours) recommended for the management of patients with heterogeneous, disabling chronic pain in a pain management program (Guzman, et al., 2001). While it may appear at first sight that there is a degree of inconsistency in providing an intervention incorporating pacing, where other evidence suggests pacing is an unhelpful strategy among those with chronic pain due to physically disabling conditions, this may be a matter of definition; pacing as taught in cognitive behavioural pain management programs takes a systematic approach to continuing to build up quotas of activity which it may be hypothesized is absent in what respondents would endorse as pacing in surveys of pain-related coping strategies. However, the findings from the evaluation of the programs suggested that there was merit in pursuing the use of cognitive behavioural pain management programs in the context of refractive spinal cord injury pain.

4.3.2 Cognitive restructuring

A pilot program exploring the use of a cognitive restructuring intervention targeting catastrophizing for a heterogeneous group of people with disability related chronic pain has recently been reported (Ehde & Jensen, 2004). The authors compared eight 90-minute sessions of cognitive restructuring with an education control intervention among 18 people with disability-related chronic pain (including those with amputations, spinal cord injury, cerebral palsy and multiple sclerosis). The cognitive restructuring intervention included: the role of negative cognitions; how to identify maladaptive thinking; thought-stopping and cognitive restructuring techniques; and use of reassuring self-statements. The education control intervention included pain education (underlying mechanisms and theories of pain), sleep problems in pain and common pain treatments. The authors report that nine of those who attended the first session did not return, but of the eighteen who did continue with their treatment all reported benefiting from the intervention regardless of the group attended. The preliminary results reported by the authors, describing mean pain intensity on a range of 0 to 10 before and after attendance, suggested that whereas pain intensity was unchanged in those attending the education group there was a reduction of approximately 0.2 of a standard deviation among those participating in the cognitive intervention. The authors conclude that a properly powered controlled trial would be required to establish the effectiveness of this approach, but that it was certainly feasible to provide and regarded as acceptable by at least half the patients. Although the literature on the use of psychosocial interventions in people with physically disabling conditions is limited, there is some evidence of similar therapeutic mechanisms operating in these populations as in chronic primary pain conditions (Burns,

Kubilus, Bruehl, Harden, & Lofland, 2003). In particular, the observation of the association between decreased pain catastrophising and improvements in mood and disability in those who participate in the interventions is consistent with findings in other chronic pain populations (Jensen, et al., 2011; Nicholson Perry, et al., 2010). This cognitive restructuring intervention therefore has particular promise as it targets pain catastrophizing, but requires implementation of a smaller range of treatment strategies than traditional cognitive behavioural pain management programs as described above.

4.3.3 Hypnosis

Hypnosis has also been applied to spinal cord injury related pain in a series of studies (Ehde & Jensen, 2007) using individual hypnosis treatments with 10 sessions over four weeks and daily practice. The suggestions used were reported to include imagery, changing sensations and ignoring pain, with associated post-hypnotic suggestions that a relaxed state and the ability to ignore pain will become increasingly easy. The case studies found that a sub-group of individuals with disability-related pain were able to obtain decreases in pain severity, with associated improvements in mood, sleep and general well-being in individual cases. However, in the absence of randomized controlled trials, no firm conclusions can be drawn about the effectiveness of hypnosis in this context.

In a variation of the more common cognitive behavioural interventions reported in the literature, a cognitive restructuring approach combined with self-hypnosis training was reported in people with multiple sclerosis (Jensen, et al., 2011). This intervention was intended to target pain catastrophising as well as pain intensity. When compared with either cognitive restructuring or hypnosis alone, or the control condition, the combined approach resulted in a decrease in the frequency of pain catastrophising and increase in the frequency of reassuring cognitions, as well as improved average and worst pain intensity.

4.4 Barriers to the use of psychosocial interventions

While access to specialist pain management services of any type is problematic, there are some additional reasons to think that access to psychosocial pain interventions will be particularly difficult for those with physically disabling conditions. Broadly, these include the demands of providing such services and accessibility of such services. Health professionals specialising in the provision of psychosocial interventions for chronic pain, such as clinical psychologists, tend to be limited in supply and concentrated in specialist services in major cities. The skills and expertise required to provide psychosocial pain management interventions to those with physically disabling conditions requires expertise in both pain and some of the specific aspects of the physically disabling condition which may impact upon the delivery of the intervention. This includes having an understanding of the physically disabling conditions and its associated symptoms, such as motor function or fatigue, and how these may impact upon the relevance or implementation of the strategies taught. The additional challenges to mobility from having a physically disabling conditions, as well as chronic pain, in combination with environmental barriers which must be overcome to attend a specialist pain management service reduces the chances that an individual with a chronic pain problem due to a physically disabling condition will be able to attend. The costs of living with a physically disabling condition may result in limited financial resources available to fund travel or accommodation in locations where specialist pain management services are available.

Service delivery models which are able to overcome some of these barriers are yet to be designed, and many of the interventions with a research pedigree to support them have not generalized to routine care due to the lack of support to assist with translation into routine clinical practice. The increased availability of high speed internet may permit the use of online interventions to provide at least some access to some of the components of effective psychosocial interventions for chronic pain, either for use alone or with the support of a health professional, and this may be of particular benefit to those with pain secondary to physically disabling conditions.

4.5 Summary

Despite the limited evidence available about the effective of psychosocial interventions in people with physically disabling conditions, the findings suggest that there is merit in further research to evaluate their usefulness in a broader range of conditions. Intervention studies in this area are notoriously difficult, due to the many barriers to participation and retention in trials. National and international collaborations are likely to be necessary to ensure a sufficient sample size for such studies to be adequately powered. Provision of these services is impeded by a number of practical barriers, some of which might at least partially addressed by making more effective use of information technology (World Health Organisation, 2011). While there is currently insufficient evidence to support a wholesale recommendation to use these interventions in all physically disabling conditions, in light of the dissatisfaction with pain treatment among many with chronic pain secondary to physically disabling conditions they may be considered for use on an individual basis.

5. Conclusion

It can be concluded based upon the data presented that the application of biopsychosocial models to the understanding, assessment and management of chronic pain associated with physically disabling conditions is at an early stage of development. There is a well-established body of research in some conditions, such as spinal cord injury, whereas almost nothing is known about the application of these models to other conditions, notably Parkinson's Disease. Many of the patterns observed in primary pain conditions are replicated in these conditions, but the exceptions noted underscore the importance of caution in generalising findings from one condition to another. Relatively, research concerning the use of psychosocial interventions is less well-developed than research examining the relationships between psychosocial and pain variables in physically disabling conditions. Despite this, the findings generally are suggestive of an important role of including psychosocial variables in our conceptualization of individual differences in the experience of chronic pain and its consequences in people with physically disabling conditions, and the possibility of improved outcomes through the use of psychosocial interventions.

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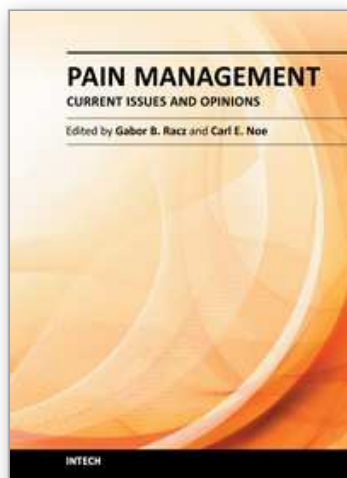
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